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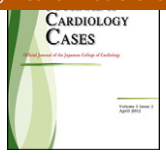

Case Report

Perforation of mitral-aortic intervalvular fibrosa secondary to bicuspid aortic valve endocarditis: Possible relation with diagnostic coronary angiography

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ABSTRACT

Infective endocarditis (IE) remains a serious and deadly disease despite recent advances in diagnosis and treatment. In all IE cases, the rate of healthcare-associated IE has been reported as 23%. Aortic valve endocarditis may extend to mitral-aortic intervalvular fibrosa (MAIVF), which may cause pseudoaneurysm formation and subsequent perforation. Direct perforation of the MAIVF is a rare clinical finding. In this report, we present a case of bicuspid aortic valve endocarditis which manifested as acute heart failure secondary to perforation of MAIVF and developed after diagnostic coronary angiography.
<**Learning objective:** Direct perforation of MAIVF without abscess or aneurysm formation is a rare complication of IE. Health-care associated IE (HAIE) occurs mostly secondary to vascular manipulations and coronary angiography (CAG) is a rare cause of HAIE. Here, we report a case of aortic valve IE which developed two weeks after CAG and was complicated with perforation of MAIVF. *Staphylococcus epidermidis* was the causative microorganism. Urgent surgical treatment resulted in complete recovery of the patient.>

Introduction

Healthcare-associated infective endocarditis (HAIE) is defined as either IE manifesting >48 h after admission to hospital or IE acquired in association with a significant invasive procedure performed in the 6 months before diagnosis [1]. Vascular manipulation is the leading source of bacteremia causing HAIE [2]. Native aortic valve may be affected in HAIE and peri-annular extension of the infection may cause serious complications including pseudoaneurysm formation and perforation of mitral-aortic intervalvular fibrosa (MAIVF). In this report, we present a case of HAIE complicated with perforation of MAIVF which developed after diagnostic coronary angiography (CAG).

Case report

A 54-year-old male patient was admitted to the emergency department with complaints of fever and dyspnea. His medical history revealed that 1 month previously, severe regurgitation of bicuspid aortic valve, left ventricular dilatation, and systolic dysfunction were diagnosed, operation was recommended, and pre-operative CAG was performed due to accompanying angina pectoris. Using modified Seldinger technique, a 6-French sheath was placed in the right femoral artery and selective visualization of coronary arteries was performed with left and right Judkins catheters. The patient tolerated the procedure well and no complication involving femoral artery and puncture site was encountered. Critical stenosis of the right coronary artery was established and the patient was scheduled for an elective aortic valve replacement and coronary artery bypass grafting operation. Two weeks after CAG, he developed fever and malaise that progressed to severe dyspnea within 2 days. On physical examination, he had a fever of 39 °C, heart rate 130 beats/min, blood pressure 100/65 mmHg, and respiratory rate 25 breaths/min. On heart auscultation, a diastolic murmur along the left sternal border and a grade 2/6 pansystolic murmur at the apex were recognized. His lung examination revealed bibasilar rales. Laboratory data showed elevated inflammation markers with a white blood cell count of 15,000/μL, sedimentation rate of 54 mm/h, and C-reactive protein of 19 mg/L.

Parasternal long-axis and short-axis views on transthoracic color Doppler echocardiography showed a turbulant flow between left ventricular outflow tract (LVOT) and left atrium (LA) (Fig. 1). In apical 4-chamber view, a fistula located at the junction of anterior mitral leaflet and non-coronary aortic valve cusp was seen. Multiple echogenic masses consistent with vegetations were attached to the atrial site of the fistula and there were no vegetations on the bicuspid aortic valve. An echo-free space around the annulus compatible with pseudoaneurysm or abscess formation was not

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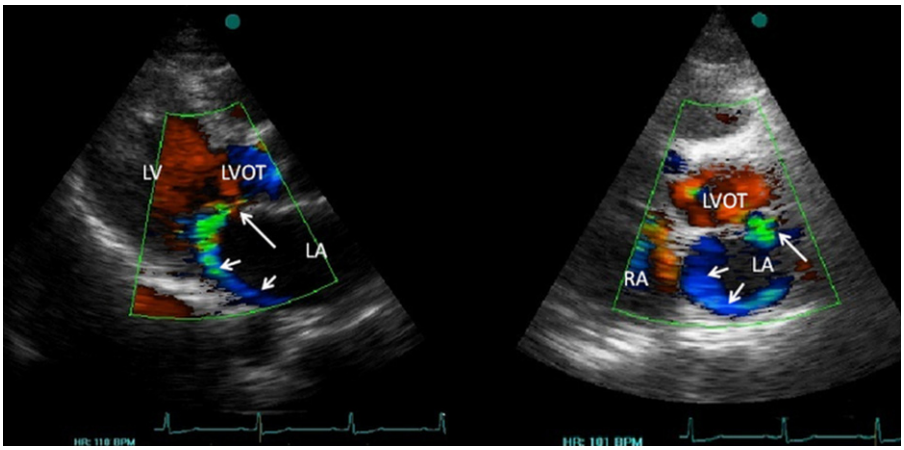

Fig. 1. Transthoracic Doppler echocardiography of LVOT and LA in parasternal long-axis view (left panel) and short-axis view (right panel) showing eccentric turbulent flow originating from MAIVF, propagating parallel to mitral annulus and along the posterior LA wall (arrows). LA, left atrium; LV, left ventricle; LVOT, left ventricular outflow tract; MAIVF, mitral-aortic intervalvular fibrosa; RA, right atrium.

observed (Fig. 2). Continuous wave (CW) Doppler tracing of the fistula showed that the flow between LVOT and LA have systolic and diastolic components (Fig. 2). Unfortunately, transesophageal echocardiography could not be performed due to the hemodynamic instability of the patient. The findings were compatible with periannular extension of aortic valve IE which was complicated with perforation of MAIVF.

Three blood cultures were taken with 1 h duration in between, empirical antibiotic therapy was initiated, and urgent cardiac surgery was performed. Briefly, saphenous vein graft was anastomosed to right coronary artery, infected aortic valve was resected, the vegetations on the atrial site were debrided, the defect between LVOT and LA (Fig. 3) was closed with a pericardial patch and finally No. 23 mechanical heart valve was implanted to the aortic position.

Postoperative course of the patient was uncomplicated. Control transthoracic echocardiography showed complete closure of the defect and normal functioning prosthetic aortic valve. Three blood cultures and culture of the resected valve revealed *Staphylococcus epidermidis* as the causative microorganism. The patient was discharged after 6 weeks of antibiotic therapy.

Discussion

The clinical appearance and microbiological causes of IE have changed since the first description and the incidence of HAIE has increased significantly reaching up to 23% of IE cases [3]. HAIE has a higher mortality and morbidity rate when compared to community acquired IE. Risk of HAIE is relatively high in elderly patients with renal failure, diabetes mellitus, and degenerative valvular disease who have vascular manipulations including hemodialysis, receipt of chemotherapy, and coronary interventions. Prompt diagnosis and initiation of antibiotic therapy combined with surgical intervention is necessary to reduce the mortality rates.

The risk of endocarditis associated with catheter-related bacteremia in patients with long-term central venous catheters is well defined. However, other vascular manipulations including CAG and percutaneous coronary intervention are widely performed in the ambulatory setting and may also induce bacteremia. The incidence of bacteremia secondary to cardiac catheterization procedures has been reported as 0.4–0.6% and in 5.5% of HAIE cases CAG was the source of infection [4,5].

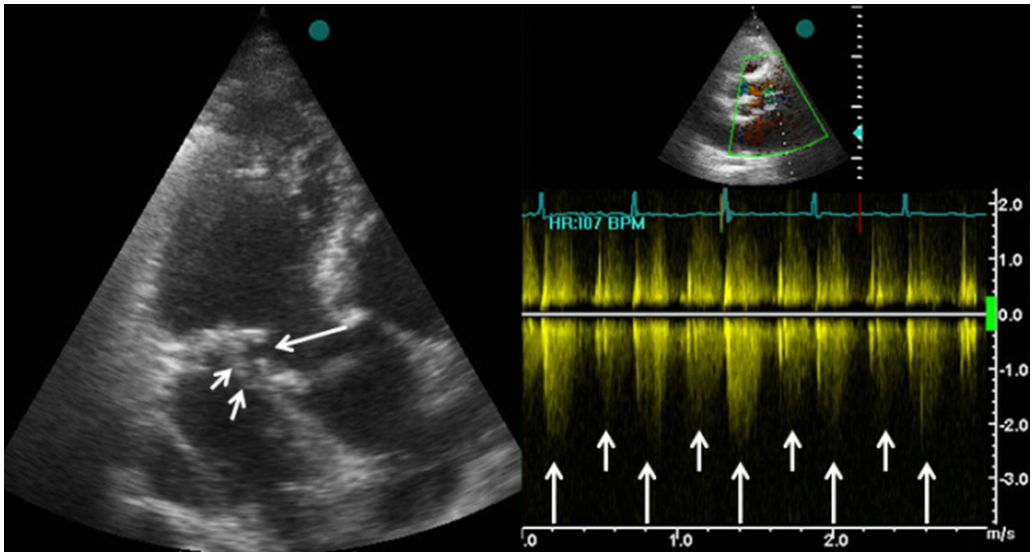

Fig. 2. Transthoracic echocardiography apical 5-chamber view showing perforation of mitral-aortic intervalvular fibrosa and multiple echogenic masses consistent with vegetations adherent to the atrial site of the fistula (left panel). Continuous wave Doppler tracing shows systolic (long arrows) and diastolic (short arrows) flow from the left ventricular outflow tract to the left atrium through the fistula (right panel).

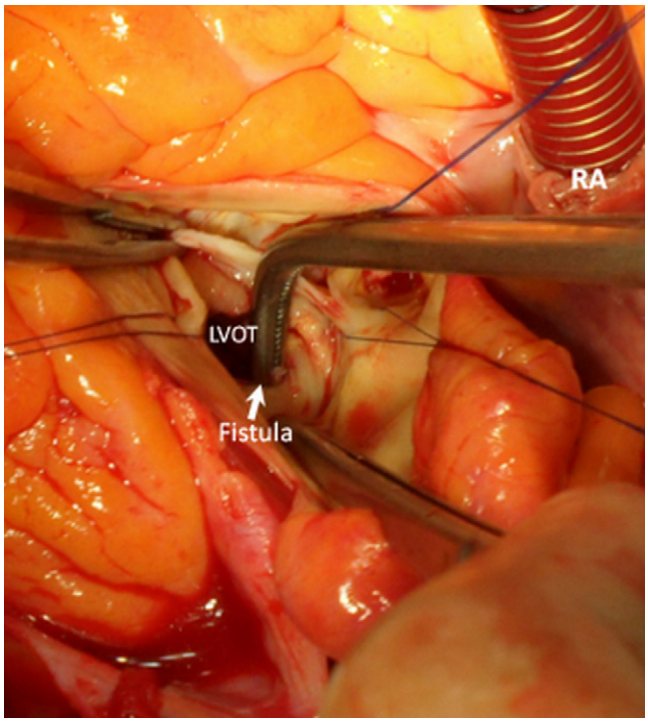

Fig. 3. Intra-operative view of the patient through the aortotomy. Fistula located on the posterior aspect of left ventricular outflow tract, at the junction of the non-coronary cusp of aortic valve and anterior mitral leaflet is indicated with the dissector (arrow). RA, right atrium.

Staphylococcus species are the most causative agents causing 31.2% of all IE cases and 70% of HAIE [3]. Coagulase negative staphylococci including *S. epidermidis* cause 7.8% of native valve endocarditis and may directly infect the valvular annulus leading to valve destruction and heart failure [6]. Monk et al. reported that *S. epidermidis* isolates obtained from patients with native valve endocarditis are more virulent when compared to isolates obtained from patients with prosthetic valve endocarditis [7]. In our case, the cultures of blood and operation samples were positive for *S. epidermidis*. In addition, the only possible source of infection was contamination from the skin during the puncture of femoral artery in the CAG procedure. According to these findings, we concluded that the diagnosis in our patient was HAIE secondary to CAG.

Peri-annular extension of the infection may result in severe complications including abscess or pseudoaneurysm formation. Sudhakar et al. reviewed 89 cases with aortic pseudoaneurysm most of which occurred in prosthetic valve endocarditis [8]. Direct perforation of the MAIVF without pseudoaneurysm formation in native aortic valve endocarditis is very rare and should be differentiated from perforation of anterior mitral leaflet and aneurysm of sinus valsalva [9,10]. Eccentric aortic regurgitation jets striking to the MAIVF may facilitate inoculation of the microorganisms to the injured endocardium and may play a role in perforation of the MAIVF in the early stages of the disease before abscess or pseudoaneurysm formation. Unusual echocardiographic findings of this kind of defect have been described in detail in a report by Konka et al. [11]. Similar to the previous data, in CW Doppler tracing we observed a systolo-diastolic flow through the perforated segment which included (1) a systolic flow during the ejection phase of blood through the LVOT and (2) aortic regurgitation flow entering the LA. In our opinion, the unusual mechanics of the blood flow through the fistula may explain the rapid progression of heart failure symptoms due to abrupt increase in LA pressure. In IE, urgent surgical intervention is recommended for patients with peri-annular extension of infection.

To conclude, perforation of MAIVF may occur without accompanying peri-annular complications like pseudoaneurysm or abscess formation in IE caused by *S. epidermidis*. Even though current guidelines do not recommend IE prophylaxis before coronary angiography, strict antiseptic precautions must be taken to reduce the risk even more in patients with risk factors for HAIE.

Acknowledgment

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